J Ped Surg Case Reports 11 (2016) 14–16

Contents lists available at ScienceDirect

Journal of Pediatric Surgery CASE REPORTS

journal homepage: www.jpscasereports.com

Latent vitello-intestinal duct in an infant: Unique presentation with trans-umbilical retrograde post-operative intussusception following cardiac surgery

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ARTICLE INFO

Article history:
Received 26 April 2016
Received in revised form
17 May 2016
Accepted 19 May 2016

Key words:
Vitello-intestinal duct
Ileal prolapse
Transposition of Great Arteries
Intussusception
Post-operative
Retrograde
Umbilical

ABSTRACT

Persistent vitello-intestinal duct (PVID) is a common gastro-intestinal anomaly seen in neonates. There have been a few case reports of the distal ileal loop herniating out of the PVID. However, all these occurrences have been noted in the immediate post-natal period. We present here an interesting case of latent vitello-intestinal duct presenting as a trans-umbilical retrograde post-operative intussusception in a 2 month old infant after undergoing an open corrective cardiac surgery for Transposition of Great Arteries (TGA).

1. Introduction

Incidence of PVID is commoner than reported and the usual symptom is umbilical discharge or an umbilical granuloma [1]. Most of these patients are diagnosed at birth or within the first month of life and undergo corrective surgery. This holds true for patent VID and not necessarily for other sequal of VID like a cyst, sinus or Meckel's diverticulum. We present here a case of a previously asymptomatic 2 month old child who presented with intussusception of the distal ileal loop through the PVID 8 days after undergoing a major cardiac surgery.

2. Case history

A 2 month old first born male child, diagnosed case of Transposition of Great Arteries (TGA) underwent corrective surgery for the same at our institute. Patient was otherwise asymptomatic and had no other history of any illness apart from the cardiac condition. He was on full feeds prior to surgery and resumed full feeds on postoperative day 6 though cardiac pacing was being done by a pacemaker, as there was no spontaneous rhythm till then. On postoperative day 8, sudden herniation of the bowel loops was noted from the umbilicus. Patient underwent an emergency exploratory laparotomy where it was discovered that the herniated bowel loop was actually the distal ileal segment which had intussuscepted through the patent vitello-intestinal duct (Fig. 1). The intussusception was reduced manually and was viable (Fig. 2). Following reduction of the intussusception, it was apparent that it was a retrograde one originating from the distal limb of the ileum all the way through a persistent vitello-intestinal duct (Fig. 3).

Wedge excision of the VID was done and bowel continuity restored.

Post-operative course was uneventful. External pacing was discontinued on postoperative day 4 and full feeds resumed on post-operative day 5. Patient made an uneventful recovery and is thriving well on follow up.

3. Discussion

Persistent vitello-intestinal duct (PVID) may be patent, latent or initially patent followed by closure due to either the acid secretion from the ectopic gastric mucosa or iatrogenic chemical or electrical

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http://dx.doi.org/10.1016/j.epsc.2016.05.008

Fig. 1. The herniated intussuscepted distal ileal loop.

Fig. 3. Patent VID.

cauterization. PVID may manifest with prograde (through proximal limb), retrograde (through distal limb) or orthograde intussusception involving both limbs in cases of very short and wide PVID. Post-operative intussusception following excision of PVID has been reported but none for the trans-umbilical presentation of PVID in the post-operative period [2].

Persistent vitello-intestinal duct is a common surgical problem in neonates. Its various iterations and presentations are well documented in literature [1–5]. A PVID generally presents as a discharging umbilical sinus or an umbilical granuloma. Dictum is that every umbilical granuloma should be investigated for the presence of a PVID by means of an ultrasound examination [4].

The incidence of bowel or mucosal prolapse through the PVID, though a rare event, is well documented in literature [6–8]. Numerous reports are found in literature elucidating the prolapse of the distal ileal loop or the mucosa through the PVID. Most presentations are in the immediate post-natal period or the neonatal age group. Late and unusual presentations of PVID are known to occur [9,10]. The association of PVID with mucosal prolapse with omphalocele is also well documented [11–16].

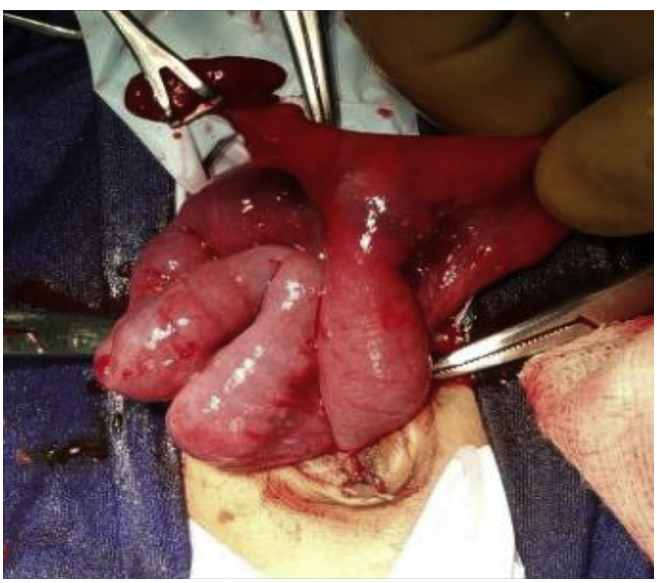

Fig. 2. The intussusception being reduced.

But late presentation of latent VID with the intussusception of the distal ileal loop as its presenting feature is unique. The patient was an asymptomatic infant on full feeds awaiting corrective surgery for the cardiac anomaly with no symptoms pertaining to the bowel anomaly in particular.

Not only was the presentation sudden and unexpected but was of an unusual nature as well.

Diligent history and examination though being the pillars of diagnosis in PVID, it can still present in an unusual as well as a delayed presentation.

A search for relevant literature produced only four articles that described the presence of a PVID with presence of a mucosal prolapse [11–16]. A review of these articles indicated a common pattern that this anomaly was picked up very early in neonatal life. Most of the patients were symptomatic even prior to the presentation with symptoms ranging from an evidence of umbilical granuloma, watery discharge from the umbilicus or feculent discharge from the umbilicus. Review of the case reports about the PVID per se also indicated presence of some symptomatology at the umbilicus from the time of birth till the time of presentation or diagnosis.

In our patient, there was no evidence of any symptom prior to the presentation. A detailed history with direct questioning was elucidated from the parents after the surgery but there was no indication regarding the presence of any umbilical pathology.

Patient was diagnosed in the antenatal period to be suffering from a congenital cardiac disease and was being monitored and worked for it. This presentation of PVID with or without the presence of a mucosal prolapse and at this late stage is uncommon and not reported in literature. This further reiterates our belief that congenital anomalies in children rare or common can present in an uncommon and atypical way and one can never be too careful.

Prenatal diagnosis of PVID is difficult but high alpha fetoproteins in the absence of other anomalies may raise the suspicion. Neonatal screening examination will miss it as the umbilical cord is still attached unless there is some discharge or stains. An examination

of the umbilicus later on or examination under anesthesia when undergoing any other surgery such as this patient may manifest and occult pathology. High index of suspicion leading to careful introduction of probe, ultrasound examination or contract study may finally unravel it [17].

Management options may include reduction of intussusception under analgesia, sedation or anesthesia followed by trans-umbilical exploration and definitive surgery or laparoscopic or open laparotomy would help. Wedge resection in a viable case without any evidence of ectopic tissue would suffice. Resection may be required in cases of ectopic tissue, non-viable bowel any complications of gangrene or perforation [18].

Conflict of interest

Nil.

Source of funding

Nil.

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